

# ADENOID CYSTIC CARCINOMA OF BARTHOLIN'S GLAND: A CASE REPORT

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## SUMMARY

**Objective:** Adenoid cystic carcinoma (ACC) of Bartholin's gland is a distinctive entity of vulvar malignancy, accounting for 10–15% of Bartholin's gland carcinomas. The purpose of this paper is to report our diagnosis and surgical treatment of a case of primary Bartholin's gland carcinoma presenting as a Bartholin's cyst.

**Case Report:** A 49-year-old woman was referred due to the increasing size of a posterior vulvar lump despite aspiration. Histology of the biopsy specimen confirmed ACC of Bartholin's gland. The patient was treated using radical vulvectomy with inguinal lymph node dissection. No evidence of local recurrence or distant metastasis was noted up to 18 months after surgery.

**Conclusion:** Although uncommon, Bartholin's gland carcinoma should be suspected when excess induration or necrotic debris is present or when a Bartholin's cyst or abscess is unresponsive to conventional therapy, especially in perimenopausal and postmenopausal women. [*Taiwanese J Obstet Gynecol* 2005;44(1):72–74]

**Key Words:** adenoid cystic carcinoma, Bartholin's gland

## Introduction

Primary carcinoma of Bartholin's gland is a rare malignant tumor arising from the posterior half of the vulvar region. Primary adenocarcinoma of Bartholin's gland represents less than 1% of all genital malignancies and 2–7% of all vulvar carcinomas. Adenoid cystic carcinoma (ACC) of Bartholin's gland is a distinctive entity of vulvar malignancy, accounting for 10–15% of Bartholin's gland carcinomas [1]. The clinical course of ACC is protracted, with a tendency for perineural and local invasion [2]. Distant metastasis may appear several months or years after local recurrence.

## Case Report

A 49-year-old woman, gravida 4, para 3, presented with a painless lump in her left labial area for about 2 years. Initially, she consulted a local gynecologist and a Bartholin's cyst was diagnosed. Aspiration was performed but the mass persisted. She was referred to our hospital in August 2002 due to the increasing size of the lump.

The lump was incised and drained. Histology of the biopsy specimen confirmed ACC of Bartholin's gland. The patient underwent radical vulvectomy with inguinal lymph node dissection. The surgical specimen revealed the same histologic findings as the biopsy. The surgical margins and bilateral inguinal lymph nodes were free of tumor. The postoperative course was uneventful. The patient has been under observation for 18 months without any recurrence or metastasis.

## Discussion

Bartholin's gland is located deep in the bulbospongiosus muscle, adjacent to the introitus. The diagnostic criteria for primary Bartholin's gland carcinoma must include

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anatomic position in the Bartholin's gland with intact overlying skin. This is an adaptation of the stricter criteria of Honan [3].

Adenocarcinoma arising from the mucin-secreting columnar epithelial cells of the acini accounts for approximately 40% of Bartholin's gland carcinoma. Squamous cell carcinoma arising from the squamous epithelium at the vestibular orifice accounts for another 40% [4]. Other types include ACC (10–15%), transitional cell carcinoma (< 5%), poorly differentiated adenocarcinoma, and clear cell carcinoma [5,6].

The age at presentation with Bartholin's gland carcinoma is 50–60 years. The tumor generally presents as a painless lump in the posterior part of the labium major [7,8]. Other symptoms include bleeding, pruritus, and pain. The reported duration of presenting symptoms ranges from 3 weeks to 17 months [9,10]. The interval from the onset of symptoms to definite diagnosis is not correlated with tumor size, stage of disease, or outcome [9]. Most patients are initially diagnosed with Bartholin's cyst or abscess and are treated with incision and drainage or marsupialization. Failure of resolution of the mass ultimately leads to biopsy and appropriate diagnosis.

The extreme rarity of Bartholin's gland carcinoma makes it difficult to perform large studies to define the optimal treatment. Various therapeutic modalities have been suggested, including wide excision, radical hemivulvectomy, radical vulvectomy, and pelvic exenteration. Whether unilateral or bilateral inguinal lymphadenectomy is necessary is controversial. Surgical resection with adequate margins may disrupt the function of adjacent organs. Morbidity after such radical procedures is considerable, although reconstruction may be possible [11,12]. Chemoradiation plus conservative surgery has been proposed as a therapeutic alternative to extensive radical surgery [9,11–14].

ACC of Bartholin's gland is similar to ACCs derived from salivary glands, the upper respiratory tract, and breast. The most common type of ACC of Bartholin's gland has a cribriform pattern, characterized by nests containing hyalinized basement membrane-like material [4]. Some ACCs show a tubular pattern of growth, whereas others are mainly solid. The pattern of growth is used as a grading system in salivary glands [15]. The tubular pattern is regarded as the best differentiated and the type with most favorable prognosis [16]. The solid type is associated with a higher incidence of metastasis and a significantly lower survival rate.

Local recurrence can be expected even in the case of negative surgical margins. Copeland et al [17] and Lelle et al [18] have reported 10-year progression-free intervals of 38% and 33%, respectively, compared to

survival rates of 59% and 100%, respectively. This confirms a protracted, although malignant, clinical course with this disease entity.

Although uncommon, Bartholin's gland carcinoma should be suspected when excess induration or necrotic debris is present and when a Bartholin's cyst or abscess is unresponsive to conventional therapy, especially in perimenopausal and postmenopausal women.

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